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Case Report

Uncommon presentation of cutaneous leishmaniasis: late-onset facial involvement after a decade—a rare case report

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Abstract

Leishmaniasis is a parasitic infection that can involve the skin, mucosal membranes, and internal organs. It is endemic to the tropics. A forty-three-year-old male, diagnosed and treated for visceral leishmaniasis 15 years ago, presented with a complaint of a progressively growing lesion on his face for five years. A detailed history, clinical examination, and histopathological examination were done to reach a diagnosis of cutaneous leishmaniasis (CL). Treatment with oral fluconazole 300 mg daily for six months, followed by 50 mg miltefosine three times a day for another two months, led to complete resolution of symptoms, and the lesion on the face was cured. Early diagnosis is crucial to averting unwarranted treatment and potential complications. This case aims to inform clinicians of the high index of suspicion when diagnosing CL; given the delayed presentation, the previous medical history and the natural course of the lesion are of utmost importance.

INTRODUCTION

Leishmaniasis is the result of infection with the intracellular protozoa *Leishmania donovani,* which is transmitted by infected female sandflies [1]. Cutaneous leishmaniasis (CL) is increasingly prevalent worldwide, and while it rarely leads to fatalities, it falls under the category of neglected diseases. There are about 1.5 million new cases of CL each year, of which more than 90% occur in Middle Eastern countries like Afghanistan, Algeria, Iran, Iraq, and Saudi Arabia in the old world and Brazil and Peru in the new world. The severity of cutaneous leishmaniasis is related to a variety of risk factors, such as poverty, malnutrition, migration, and poor housing conditions. If the infection of cutaneous leishmaniasis lasts for more than two years, then it is known as chronic cutaneous leishmaniasis [2]. Because it has many clinical presentations, it can mimic many dermatoses, most commonly cutaneous cancers, discoid lupus erythematosus, lupus vulgaris, and sarcoidosis, and can lead to misdiagnosis and unnecessary treatment in endemic areas [2–4]. To our knowledge, this case highlights the most temporally distant reactivation after a 10-year period.

CASE REPORT

A 43-year-old man, a trekking guide by profession, visited the office with complaints of a mildly itchy red mass over the upper lip area for five years (Fig. 1). He was apparently well five years ago and had been working in Dubai as a security guard when he noticed a small acne-like lesion in the upper lip area, which gradually increased in size and became purulent. Treatment was done in a health center in Dubai and showed no clinical improvement. So he came back to Nepal and visited multiple centers, where a biopsy was done, a diagnosis of cutaneous tuberculosis was made, and he was treated with anti-tubercular therapy for nine months.

Given the lack of therapeutic effect, the lesion rather increased in size and became more crusted.

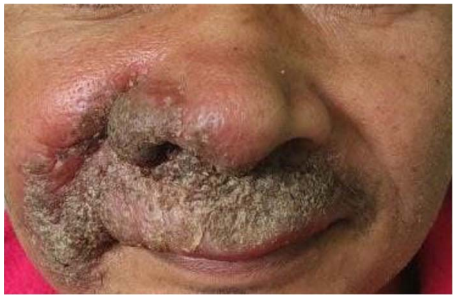

Figure 1. Erythematous plaques on upper lip area and right cheek area with crusts.

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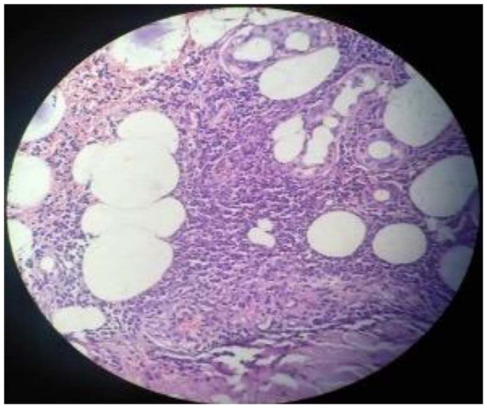

Figure 2. Histopathology showing dermis densely infiltrated with lymphocytes, histiocytes and plasma cells (magnification 10×).

Figure 3. Partially healed plaque after taking oral fluconazole for three months.

Figure 4. Complete resolution of the plaque after taking miltefosine for two months.

He then visited our center, and examination revealed a well-defined erythematous plaque of size 5 × 4 cm on the upper lip area, extending to the right nose and right cheek, with overlying crusting and surrounding erythematous areas. The mass was non-tender on palpation, without associated lymphadenopathy. The sensations over the lesion were intact, which ruled out leprosy.

The biopsy was repeated, but the report was inconclusive. The report showed the dermis densely infiltrated with lymphocytes, plasma cells, and histiocytes. There were vague aggregates of epithelioid histiocytes. Amastigotes of Leishmania species were not found. The epidermis was thin. The Periodic acid–Schiff (PAS) stain was negative for fungal elements, and the Ziehl–Neelsen (ZN) staining was also negative for acid-fast bacilli (Fig. 2).

In the past 15 years, he has suffered from visceral leishmaniasis (VL) and was admitted to a tertiary hospital in eastern Nepal. Treatment was started in line with cutaneous leishmaniasis with oral fluconazole 300 mg per day for six months. The lesion size gradually decreased within three months of starting oral fluconazole (Fig. 3). Routine lab tests were done, which were all normal, and later liver function test monitoring was done every month. To rule out syphilis, the rapid plasma reagin (*RPR*) titer was sent, which was negative. Though there was some improvement, it was not the anticipated improvement sought within the span of six months. So, the medication was changed to oral miltefosine 50 mg three times a day for 28 days. Within two months of initiating oral miltefosine, a significant improvement was observed, and the lesion disappeared completely (Fig. 4).

DISCUSSION

The manifestations of CL are broad and may mimic other inflammatory and neoplastic diseases. Leishmaniasis is a polymorphic parasitic infection caused by an obligate parasite of the genus Leishmania. Leishmaniasis presents in three forms: VL, mucocutaneous (MCL), and CL, with CL being the most common form. Although not as severe as the MCL or VL, the consequences of the disease can be severe, including permanent scarring, disfigurement, possible dissemination of the parasite, and an increased risk for skin cancer. Instances of delayed manifestation of cutaneous leishmaniasis, as in our case, have been rarely reported in the literature [5, 6].

Clinically, a typical CL lesion first appears as a small red papule, which is commonly mistaken for an MRSA infection or furuncle. Exposed body areas such as the face or extremities are the most common locations, which are unfortunately also common areas where malignant neoplasms such as basal cell carcinoma and squamous cell carcinoma can occur. Within 1–3 months, the infection may progress into erythematous nodules, indurated or scaly plaques, or ulcers with raised borders. As this is a case of chronic cutaneous leishmaniasis, there were no amastigotes, but a large number of plasma cells, lymphocytes, and histiocytes were present. The lesions are pruritic, generally not painful, and unresponsive to therapy with antibiotics or corticosteroids. Systemic symptoms are often absent but have been reported. Typically, lesions heal spontaneously between six months and three years. The diagnosis of CL is critical, both to avoid unnecessary treatment such as surgical intervention and to initiate proper therapy to reduce the risk of disease complications.

Although the specific factors contributing to late reactions can vary across individuals, immunosuppression and persistent parasite reservoirs have been deemed significant. The chronicity, persistence of lesions, and delayed or inadequate clinical response to treatment are attributed to the Th2-type immune response [7]. Cutaneous leishmaniasis may present with many clinical entities. We documented a case of CL from an endemic region, mimicking cutaneous tuberculosis in Nepal, and initially misdiagnosed and treated likewise. The consequence of a missed diagnosis is unnecessary exposure to toxic anti-tubercular therapy, which later on may lead to drug resistance. Even a delay in diagnosis may lead to disseminated disease, scarring, and the spread of infection. Scarring may be associated with decreased quality of

life, stigmatization, and long-term psychological consequences. Relying solely on histopathology may lead to an underdiagnosis of the disease, as leishmaniasis shares many features with other granulomatous diseases. A PCR for leishmania-specific DNA must be performed in any unusual granulomatous dermatitis, and this test may serve as a link between clinical and histopathological presentation. It has high sensitivity (97%–100%) regardless of the age of the lesion. Prevention and control require a multipronged approach that includes vector control, disease surveillance, timely diagnosis, and appropriate treatment. In our case, oral fluconazole and miltefosine were used, which led to complete remission of the lesion within eight months. Antimony-based compounds like N-methylglucamine antimoniate and sodium stibogluconate, via either the local or systemic route, are frequently utilized for treatment. Additionally, pentamidines and amphotericin B have demonstrated clinical effectiveness [4, 8].

Although facial involvement is a common presentation, the chronological latency between primary infection and reactivation is considered atypical. The aim of this report is to extend awareness of the atypical presentation of cutaneous leishmaniasis and emphasize the importance of considering the possibility even in cases with a prolonged time interval between exposure and symptoms. To our knowledge, this case highlights the most temporally distant reactivation.

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CONFLICT OF INTEREST STATEMENT

No conflicts of interest.

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ETHICAL APPROVAL

The Institutional Review Board deemed the study exempt from review.

CONSENT

Written informed consent was obtained from the patient.

GUARANTOR

Amrita Shrestha.

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